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On the Structure of Motor Symptoms of Parkinson's Disease

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Abstract: **This study aims to investigate the structure of the motor symptoms of Parkinson's disease (PD), as measured by the Motor Section of the Unified Parkinson's Disease Rating Scale (UPDRS). The dimensionality of the Motor Section of the UPDRS was studied using structural equation modeling. The UPDRS measures were obtained from 405 patients with PD [237 men (39 "off", 170 "on", 28 unknown) and 168 women (21 "off", 140 "on", 7 unknown)]. The ordinal character of UPDRS scores and sample size substantiated the use of robust diagonally weighted least squares model estimation. It was shown that the Motor Section of the UPDRS incorporates five main latent symptom factors (rigidity, tremor, bradykinesia of the extremities, axial/gait bradykinesia, speech/hypomimia) plus two additional factors for laterality, which account for asymmetry of tremor, rigidity and bradykinesia of the extremities. Tremor seems to be an independent symptom factor of PD. Other latent variables are substantially correlated.** © 2008 Movement Disorder Society

Key words: Parkinson's disease; structural equation modeling; dimensionality; Motor Section of the UPDRS

The identification of symptom groups of neurological syndromes such as the combination of hypokinesia, rigidity, resting tremor, and postural abnormalities in Parkinson's disease (PD) is important because knowledge about the co-occurrence of symptoms may help to define disease phenotypes and provide clues for differential diagnosis. The number of symptom groups (dimensionality) can be inferred through statistical analysis of measurements used for impairment evaluation. Within the Motor Section of the Unified Parkinson's Disease Rating Scale (UPDRS), main motor symptoms of PD (tremor, rigidity and bradykinesia) and axial symptoms (speech, posture, postural stability and gait) define symptom groups which are in practice evaluated regarding their respective severity. This paper discusses the dimensionality of the Motor Section of the UPDRS and the structure of motor symptoms of PD within the framework of structural equation modeling (SEM) using confirmatory factor analysis.

In previous studies on dimensionality assessment of the Motor Section of the UPDRS,[1–4] between three and six factors were found with percentages of explained total scale variance ranging between 59% and 78%. All these studies used exploratory factor analysis (EFA) methods, principal component analysis included. Such procedures rely on strong assumptions concerning either the distribution of observed variables, their level of measurement, or the number of observations. Principal component analysis requires a continuous measurement level[5,6]; maximum likelihood estimation in EFA requires continuous measurement levels and either normally distributed item responses or a large number of observations which may compensate for small degrees of nonnormality.[7,8] Given the ordinal distributional properties of the items in the Motor Section of the UPDRS, previous conclusions on dimensionality may not be trustworthy because the validity of assumptions of EFA modeling is lacking.

Instead of EFA, we used confirmatory factor analysis (CFA) within a SEM framework to perform a statistical test and to evaluate a number of plausible factor models for the structure of symptoms underlying the Motor Section of the UPDRS. Some SEM estimators are designed for ordinal measurements and thus, in principle, suited for analyzing that structure.

PATIENTS AND METHODS

Sample

The study includes 405 consecutive patients (237 men, 168 women, mean age 61, range 35–80 years) with PD diagnosed according to current clinical criteria.[9] Each patient was evaluated by one member of a group of certified neurologists, movement disorder specialists who routinely use the UPDRS. Sixty patients were examined in defined "off" state, and 310 patients in defined "on" state. For 35 patients, the motor state during evaluation was not specified.

This data consists of two subsamples. The first subsample of size N = 147 [96 men (38 "off", 30 "on", 28 unknown) and 51 women (15 "off", 29 "on", 7

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unknown)] was obtained at the Movement Disorder Centre, Charles University, Prague, Czech Republic. The second of size N = 258 [141 men (1 "off", 140 "on") and 117 women (6 "off", 111 "on")] was acquired at the University Medical Centre Groningen in The Netherlands.

Methods

For analyzing the latent structure of the 27 items of the Motor Section of the UPDRS, the LISREL program[10] was used. If the level of measurement is ordinal and sample size relatively small, as in our case, Jöreskog and Sörbom[11] recommend analyzing the matrix of estimated polychoric correlations of the observed variables along with the estimated matrix of asymptotic covariances of those estimated correlations, and to apply robust diagonally weighted least squares (DWLS) model estimation. The polychoric correlations and the asymptotic covariance matrix were computed using the PRELIS program.[12]

A number of theoretically meaningful models were compared. For the "final" model described here, the path diagram with standardized parameter estimates, the matrix of estimated polychoric correlations, goodness-of-fit statistics and indices, a summary of estimated standard errors of the parameter estimates, and the fitted residual matrix are reported; for details see Ref. 13.

RESULTS

The "final" model of the Motor Section of the UPDRS is shown in Figure 1. A number of theoretically plausible models were tested and compared before the model in Figure 1 was chosen as a most plausible one.[13] Following that conclusion, based on both model estimates and theoretical PD background considerations, the Motor Section of the UPDRS consists of seven factors. Five of them are substantive, each reflecting a PD motor symptom—tremor, rigidity (Rig), bradykinesia of the extremities (Brad), axial/gait bradykinesia (BBrad), and speech/hypomimia (Face). Two additional factors (Left and Right) reflect the asymmetry of tremor, rigidity, and bradykinesia of the extremities.

Although some fitted residuals (see Table 1) remained high, the fit statistics and indices suggest that this model need not to be rejected. Generally, the values of comparative fit index (CFI) and goodness of fit index (GFI) suggest a very acceptable fit, whereas root mean square error of approximation (RMSEA), standardized root mean square residual (SRMR), and normed fit index (NFI) indicate slightly less, but still acceptable model fit (see the values below Fig. 1). Values in the matrix of residual correlations are ranging from −0.40 to 0.34 (median of absolute value 0.04, standard deviation 0.07). The highest fitted residuals are those between action tremor items (right hand and left hand; 0.34), and surprisingly, between item action tremor—left hand and item Tremor—right lower extremity (−0.40). Values of factor loadings range from 0.11 to 0.92 (median 0.64, standard deviation 0.19); see Figure 1. The lowest factor loading (0.11) is for item tremor—right lower extremity as an indicator for latent factor Right; although the corresponding parameter test statistic is nonsignificant (standard error 0.17), it is theoretically meaningful to keep this parameter free. In general, values of estimated standard errors of the parameter estimates ranged from 0.02 to 0.17 (median 0.07, standard deviation 0.04). The estimated composite reliability of our model (by stratified coefficient alpha[14]) equals 0.94.

The four factors of rigidity, bradykinesia of the extremities, speech/hypomimia, and axial/gait bradykinesia are correlated, which is meaningful from a theoretical point of view. The correlations range between 0.54 and 0.85 (see Fig. 1) indicating rather substantial relationships among these symptom factors. Tremor, however, seems to be a PD symptom occurring independently of other motor PD symptom factors.

Goodness-of-Fit Statistics and Indices

- Sample size: 405
- Degrees of freedom: 300
- Satorra-Bentler's scaled χ^2 statistic: 899.33 (P = 0.0)
- Root mean square error of approximation (RMSEA): 0.070
- 90% confidence interval for RMSEA: 0.065, 0.076
- Normed fit index (NFI): 0.96
- Comparative fit index (CFI): 0.97
- Standardized root mean square residual (SRMR): 0.077
- Goodness of fit index (GFI): 0.99
- Fitted residuals: range [−0.40, 0.34], median 0.04, standard deviation 0.07.

DISCUSSION

In this study, the structure of motor symptoms of PD was investigated by applying confirmatory factor analysis models to the Motor Section of the UPDRS.

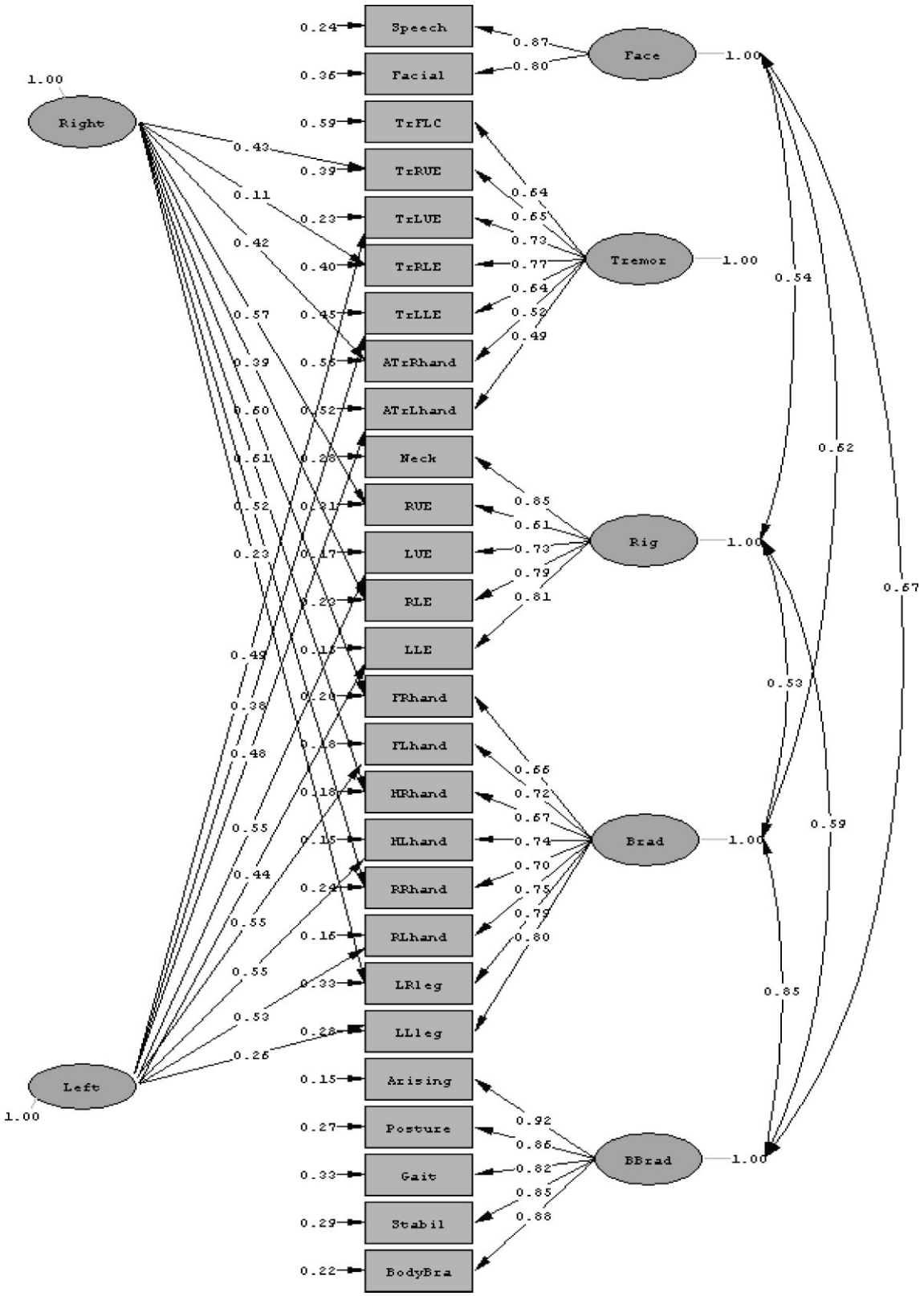

FIG. 1. Path diagram of the seven-factor model of the MS UPDRS showing estimates of completely standardized parameter estimates.

TABLE 1. *Polychoric correlations (below diagonal) and fitted residuals (above diagonal) of the Motor Section of the UPDRS*

	Speech	Facial	Lipschin	TrRUE	TrLUE	TrRLE	TrLLE	ATrRhand	ATrLhand	Rigidity	RUE	LUE	RLE	LLE	FRhand	FLhand	HRhand	HLhand	RRhand	RLhand	LRleg	LLleg	Arising	Posture	Gait	Stabil	BodyBra
Speech		0.00	0.04	−0.05	−0.05	−0.19	−0.27	−0.03	−0.02	−0.05	−0.06	−0.09	−0.01	−0.06	0.03	−0.01	0.01	−0.03	0.04	0.02	0.06	0.02	0.00	0.05	−0.03	0.00	0.00
Facial	0.70		0.19	−0.06	0.07	−0.01	−0.08	0.02	0.09	0.06	0.10	0.07	0.02	0.02	−0.04	0.00	−0.07	−0.05	−0.02	0.00	−0.04	0.02	−0.01	0.02	−0.06	−0.05	0.05
Lipschin	0.04	0.19		0.02	0.11	−0.04	−0.04	−0.21	−0.05	0.20	0.00	0.07	0.02	−0.03	0.08	0.21	0.10	0.15	0.15	0.23	0.08	0.01	0.06	0.22	0.15	0.09	0.25
TrRUE	−0.05	−0.06	0.43		0.04	0.10	−0.24	0.04	0.19	0.06	0.07	−0.01	0.01	−0.09	−0.06	−0.07	−0.07	−0.07	−0.02	−0.08	0.02	−0.13	−0.05	−0.07	0.01	−0.06	0.03
TrLUE	−0.06	0.07	0.57	0.52		−0.14	0.04	−0.13	0.00	0.13	0.00	0.05	0.05	0.02	−0.03	−0.04	−0.04	−0.07	−0.02	0.00	0.04	0.04	0.12	0.02	0.11	0.06	0.11
TrRLE	−0.19	−0.01	0.46	0.65	0.42		0.19	−0.08	−0.40	0.09	0.11	0.00	0.14	0.03	−0.07	−0.04	−0.12	−0.18	−0.03	−0.12	−0.02	−0.06	−0.03	−0.11	−0.05	−0.02	−0.01
TrLLE	−0.27	−0.08	0.37	0.18	0.69	0.68		−0.29	−0.05	0.04	−0.12	0.02	−0.07	0.10	−0.11	−0.01	−0.20	−0.12	−0.11	−0.02	0.01	0.06	−0.01	−0.01	0.03	−0.03	0.03
ATrRhand	−0.03	0.02	0.12	0.55	0.25	0.37	0.04		0.34	0.15	0.11	0.09	0.03	0.00	−0.10	−0.04	−0.07	0.03	−0.03	−0.04	0.01	−0.06	0.07	0.00	0.01	0.04	0.06
ATrLhand	−0.02	0.09	0.26	0.13	0.60	−0.02	0.45	0.60		0.19	−0.02	0.08	0.01	0.02	−0.05	−0.06	0.01	−0.01	0.01	−0.01	0.00	0.02	0.13	0.15	0.14	0.11	0.16
Rigidity	0.35	0.42	0.20	0.06	0.13	0.09	0.05	0.15	0.19		0.01	−0.03	−0.05	−0.09	0.16	0.08	0.08	0.03	0.09	0.06	0.01	0.02	−0.06	−0.01	−0.01	−0.04	0.02
RUE	0.23	0.36	0.00	0.32	0.01	0.17	−0.12	0.35	−0.02	0.53		0.09	0.05	−0.04	−0.04	−0.01	−0.07	−0.06	−0.02	−0.05	−0.02	−0.08	−0.08	−0.01	0.01	−0.03	0.04
LUE	0.25	0.38	0.07	−0.01	0.32	0.00	0.23	0.09	0.34	0.59	0.53		−0.06	−0.01	0.01	0.00	0.01	−0.03	0.01	0.00	−0.07	0.00	0.00	0.04	0.03	0.00	0.08
RLE	0.36	0.36	0.02	0.18	0.05	0.18	−0.07	0.19	0.01	0.61	0.75	0.52		0.09	−0.03	0.00	−0.07	−0.03	−0.02	−0.03	0.02	0.00	−0.07	−0.02	0.00	−0.08	0.02
LLE	0.32	0.37	−0.03	−0.09	0.23	0.03	0.26	0.00	0.23	0.60	0.46	0.82	0.73		−0.03	0.01	−0.04	−0.03	−0.03	0.00	−0.02	0.06	−0.03	0.03	0.04	−0.03	0.04
FRhand	0.38	0.28	0.08	0.20	−0.03	−0.01	−0.11	0.16	−0.05	0.46	0.51	0.27	0.48	0.26		0.11	0.05	−0.04	−0.01	−0.09	−0.02	−0.14	−0.06	−0.04	−0.01	0.00	0.05
FLhand	0.38	0.35	0.21	−0.07	0.23	−0.04	0.20	−0.04	0.21	0.41	0.22	0.58	0.30	0.56	0.59		−0.01	0.02	−0.05	−0.01	−0.12	−0.01	−0.03	−0.04	−0.07	−0.01	0.06
HRhand	0.37	0.26	0.10	0.19	−0.04	−0.05	−0.20	0.19	0.01	0.38	0.49	0.27	0.44	0.25	0.85	0.47		0.10	0.02	−0.05	−0.01	−0.15	−0.02	−0.01	0.00	−0.03	0.06
HLhand	0.37	0.32	0.15	−0.07	0.20	−0.18	0.09	0.03	0.26	0.37	0.18	0.56	0.28	0.53	0.45	0.86	0.59		−0.03	0.01	−0.10	−0.04	−0.01	0.01	−0.01	−0.01	0.06
RRhand	0.42	0.33	0.15	0.20	−0.02	0.03	−0.11	0.19	0.01	0.41	0.51	0.28	0.48	0.27	0.77	0.46	0.80	0.49		0.06	0.03	−0.10	−0.01	−0.03	0.01	−0.03	0.04
RLhand	0.42	0.38	0.23	−0.08	0.26	−0.12	0.18	−0.04	0.25	0.40	0.20	0.58	0.29	0.55	0.40	0.82	0.45	0.86	0.58		−0.09	0.00	0.04	0.01	−0.02	0.00	0.04
LRleg	0.48	0.35	0.08	0.12	0.04	0.00	0.01	0.10	0.00	0.37	0.37	0.24	0.44	0.32	0.63	0.45	0.65	0.49	0.71	0.50		0.13	0.04	−0.05	−0.05	0.00	−0.04
LLleg	0.46	0.42	0.02	−0.13	0.17	−0.06	0.16	−0.06	0.15	0.39	0.18	0.46	0.34	0.53	0.39	0.72	0.39	0.70	0.46	0.74	0.77		0.06	−0.03	−0.05	0.03	−0.03
Arising	0.54	0.49	0.06	−0.05	0.12	−0.03	−0.01	0.08	0.13	0.40	0.25	0.40	0.36	0.40	0.45	0.54	0.50	0.57	0.54	0.63	0.65	0.69		−0.01	0.03	0.05	−0.11
Posture	0.55	0.48	0.22	−0.07	0.02	−0.11	−0.01	0.00	0.15	0.42	0.30	0.40	0.37	0.44	0.43	0.48	0.47	0.54	0.48	0.55	0.52	0.55	0.78		0.05	0.02	−0.03
Gait	0.45	0.38	0.15	0.01	0.11	−0.05	0.03	0.01	0.14	0.40	0.31	0.38	0.38	0.43	0.45	0.43	0.47	0.51	0.50	0.50	0.50	0.51	0.79	0.75		0.02	−0.03
Stabil	0.49	0.41	0.09	−0.06	0.06	−0.02	−0.03	0.04	0.12	0.38	0.27	0.36	0.31	0.37	0.47	0.51	0.45	0.52	0.47	0.54	0.57	0.61	0.83	0.75	0.72		−0.07
BodyBra	0.51	0.52	0.25	0.03	0.11	−0.01	0.03	0.06	0.16	0.46	0.36	0.46	0.43	0.46	0.54	0.60	0.56	0.61	0.57	0.60	0.56	0.58	0.70	0.73	0.69	0.68	

The models were estimated using the DWLS estimator, mainly because of the ordinal measurement level of the items and the relatively small sample size.

Several studies[1–4,15] assessed the construct validity and the dimensionality of the Motor Section of the UPDRS through EFA. As discussed earlier, neither EFA nor some of the CFA estimators are the most appropriate scaling techniques, because the assumptions of the underlying statistical model may easily be violated. In previous dimensionality studies of the UPDRS, sample sizes $N < 300$ were often used.[1–3,15] In addition, measurement of the UPDRS is obviously of ordinal rather than continuous type, which may pose problems when using regular maximum likelihood estimation and PCA.[6] To our knowledge, the only study where the measurement level of the UPDRS data was respected is one by Kroonenberg et al.[16] However, their study primarily focused on the differences in the structure of PD motor signs for "on" and "off" patients; the results appeared to depend on the motor state of the patient. Their model did not fit our data, which might be due to a different scoring practice, a problem that might also account for different validity and reliability results of the UPDRS across countries.

The two factors of laterality (Left and Right) reflect the asymmetry of occurrence of tremor, rigidity, and bradykinesia of the extremities. In a clinical cohort it has been shown that initial PD symptoms start more frequently on the right-sided extremities than on the left.[17] In some EFA studies, side-sensitivity of bradykinesia of the extremities was mentioned before,[2,3] as well as that of action/postural tremor.[1] To our knowledge, side-sensitivity of rigidity and rest tremor, however, has not been previously reported.

The high correlations among the factors rigidity, bradykinesia of the extremities, axial/gait bradykinesia, and speech/hypomimia can be indicators of co-occurrence of these PD symptoms. For most patients in common PD populations, however, the main symptoms co-occur whereas isolated tremor may only be present in very early stages of PD. Further, the relative independence of tremor from rigidity and bradykinesia can be viewed as an indicator of the lack of substantive relationship between tremor and PD disability, a finding consistent with other reports.[18,19]

Since a number of theoretically meaningful models were compared, implying a partly exploratory result, future cross-validation is necessary to challenge our "final" factor structure of the Motor Section of the UPDRS. It should also be realized that larger sample sizes would make model estimation results, especially when considering the ordinal character of item responses, more reliable and final conclusions more valid.

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Psychogenic Propriospinal Myoclonus

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Video 

Abstract: **We report a case of probable psychogenic propriospinal myoclonus (PSM) in a patient who developed a sudden onset of disabling axial flexor myoclonus following a cosmetic surgical procedure. The electrophysiological findings were consistent with previous reports of PSM. Spontaneous remissions and disappearance of the jerks, sustained for 2 years, following removal of superficial surgical screws support the diagnosis of a psychogenic movement disorder.** © 2008 Movement Disorder Society

Key words: psychogenic; myoclonus; propriospinal myoclonus

Propriospinal myoclonus (PSM) is a form of spinal myoclonus characterized by involvement of muscles innervated from different segments of the spinal cord, and sequentially activated via propriospinal pathways.[1] Characteristic electrophysiological findings of slow conduction and selective recruitment of truncal and proximal limb muscles help differentiate PSM from spinal segmental myoclonus.[2] PSM has been documented secondary to intrinsic and extrinsic spinal cord lesions, and in other cases, no clear etiology has been identified. Recently the characteristic electrophysiological findings have been reported in a group of eight healthy volunteers simulating the typical axial flexor jerks of PSM.[3] The differentiation between voluntary and involuntary movements of this nature is further blurred by our report of a patient with probable psychogenic PSM.

CASE REPORT

This 65-year-old woman fell after tripping over a concrete block on the pavement, causing disfiguring soft tissue injuries above her right orbit. Apart from migraine, there were no other medical problems at the time, and no psychiatric history. There was no documented injury or pain in the neck or back following the fall, and at that time, she was neurologically normal. Legal action relating to the circumstances of the incident was initiated. A reconstructive right blepharoplasty was performed for right sided pseudoptosis. She subsequently developed a right frontal headache. The cosmetic results of the surgery were insufficient and it was revised by browplasty that required the placement of three surgical screws into the right frontal bone, including one that penetrated the frontal air sinus. The surgery was complicated by chronic pain around the operational site that was partially relieved by neck massage. Eighteen months after the fall, and following massage of the neck she developed disabling paroxysms of axial, flexor jerks that were most severe when lying supine. There was a suggestion of associated left-sided weakness at onset, but this resolved and MRI and angiogram were normal. At first the jerks occurred several times per day, but rapidly increased in frequency, with bouts of continuous jerking lasting for up to 1 hour, causing significant disability. There was positive, action myoclonus with coexistent stimulus sensitive myoclonus of variable latency, which diminished with distraction. Increasingly her mobility became affected by jerking and unsteady gait. There were periods of complete remission lasting up to several months. Jerking was exacerbated by anxiety, but no other precipitants were identified. Spinal cord and brain MRI were normal except for a few scattered deep white matter ischemic changes. Psychiatric evaluation did not identify features of somatization, depression, or malingering. She incompletely responded to piracetam 16 g per day, clonazepam 4 mg per day, sodium valproate 2 g per day, and baclofen. Three surgical screws used in the blepharoplasty were removed 4

This article includes supplementary video clips, available online at http://www.interscience.wiley.com/jpages/0885-3185/ suppmat

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